

Table 2. Characteristics of included studies – Neurologische betrokkenheid bij Myotone Dystrofie type 1 – Psychostimulantia

Study	Participants	Comparison	Follow-up	Outcomes	Comments	Risk of bias*
<i>Included in systematic review Annane, 2024</i>						
Antonini, 1997 Crossover RCT, Italy	<u>N at baseline:</u> 11 Intervention: 6 Control: 5 <u>Sex (male):</u> 55% Median age 39 years (range 21 to 61) All with myotonic dystrophy and excessive daytime sleepiness.	<u>Intervention:</u> Selegiline 20mg daily for 30 days <u>Control:</u> Placebo	4 weeks	Sleep (MSLT)	Trial was supported by a grant from Telethon-Italy	Low
MacDonald, 2002 Crossover RCT, USA	<u>N at baseline:</u> 40 <u>Sex (male):</u> 33% Mean age 41 years (SD ±12) All with myotonic dystrophy and subjective complaints of daytime hypersomnolence.	<u>Intervention:</u> 2 weeks treatment: - First week: modafinil 2dd 100mg - Second week: modafinil 2dd 200 mg <u>Control:</u> Same as intervention, yet with placebo	2 weeks	Sleep (ESS) Adverse events	Supported in part by Draxis Health Inc	Low
Orlikowski, 2009	<u>N at baseline:</u> 28 <u>Sex (male):</u> 54%	<u>Intervention:</u>	4 weeks	Sleep (MWT, MSLT and ESS)	Non-commercial funding received.	Low

Parallel group RCT, France	Aged 18 to 69 years All with genetically proven myotonic dystrophy type 1, and impairment of social or occupational functioning due to excessive sleepiness (ESS >10, MSLT ≤8).	Modafinil 300 mg daily for 1 month <u>Control:</u> Placebo 300 mg daily for 1 month		Adverse events	The study was interrupted prematurely for slow recruitment rate and drug expiration dates.	
Puymirat, 2012 Crossover RCT, Canada	<u>N at baseline:</u> 24 <u>Sex (male):</u> 50% Mean age 46 years (SD ±13) All with genetically confirmed myotonic dystrophy type 1 and excessive sleepiness (ESS >10)	<u>Intervention:</u> Methylphenidate 20mg daily at breakfast for 3 weeks <u>Control:</u> Placebo 300 mg daily for 1 month	3 weeks	Sleep (ESS and OSLER) Adverse events	Non-commercial funding received.	Some concerns (unclear risk of attrition bias)
Talbot, 2003 Crossover RCT, UK	<u>N at baseline:</u> 20 <u>Sex (male):</u> 65% Median age 43 years (range 18 to 65) All with a diagnosis of myotonic dystrophy and ESS ≥ 10	<u>Intervention:</u> Modafinil daily at breakfast: - For day 1 to 5: 100mg - For day 6 to 28: 200mg <u>Control:</u> Same as intervention, yet with placebo	3 weeks	Sleep (ESS and modified MWT) Executive function (driving, activity)	Commercial funding received with unconditional charitable grant.	Low
Wintzen, 2007	<u>N at baseline:</u> 13 <u>Sex (male):</u> 38% Mean age 43.5 years (SD ±13.9)	<u>Intervention:</u> 2 weeks treatment: - First week: modafinil 2dd 100mg	2 weeks	Sleep (ESS)	Commercial funding received with unconditional charitable grant.	Some concerns (unclear randomization, allocation concealment)

Table 2. Characteristics of included studies – Neurologische betrokkenheid bij Myotone Dystrofie type 1 – Psychostimulantia
Richtlijn Myotone Dystrofie type 1 (DM1) 2026

Crossover RCT, The Netherlands	All with myotonic dystrophy	- Second week: modafinil 2dd 200 mg or 400 mg, according to patient's perception of efficacy and tolerance <u>Control:</u> Same as intervention, yet with placebo		Apathy (interview for spontaneous activity)		
--------------------------------------	-----------------------------	---	--	---	--	--

**Assessment from the Cochrane review by [Annane \(2024\)](#)*